Opening the psychological black box in genetic counseling
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A counselee-oriented perspective on risk-communication in genetic-counseling:

explaining the inaccuracy of the counselees' risk-perception shortly after BRCA1/2-test result disclosure

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Abstract

Purpose

An important aim of genetic-counseling is helping counselees to understand the genetic contributions to their disease, such as their genetic risk to develop breast or ovarian cancer. However, many psychological studies show that their perception of their risks is often inaccurate. Previous studies showed that several information-oriented variables predict the level of accuracy, focusing on specific processes of receiving and processing risks. We choose to examine counselee-oriented predictors about how counselees embed cancer-risks in their lives. These predictors reflect the personal meaning of genetic-risks and are expected to explain/mediate the impact of genetic-counseling on risk-perception-accuracy.

Method

We analyzed 248 questionnaires of a prospective study, filled-in by probands with breast/ovarian cancer who had received pathogenic mutations, unclassified-variants or uninformative-results (resp. n=30, 16, 202). Several hypothesized predictors were used to predict the absolute level of accuracy of the counselees' risk-perception. Mediation-regression-analyses were performed to examine whether counselee-predictors mediated/explained the influence of information-predictors on the accuracy. Information-oriented predictors regarded: presentation format and communicated information, question format, education, pedigree-information, cancer experience and cognitive processes/heuristics. Counselee-oriented predictors regarded the self/personality, their life/existence in general and their need for certainty about the DNA-test, heredity and cancer.

Results

Both information-oriented and counselee-oriented variables significantly predicted the accuracy of the counselees' risk-perception, with moderate to large effect sizes. Counselee-oriented variables completely mediated/explained the effects of information-oriented variables on the accuracy.

Discussion

Counselees seem to transform the objective cancer-risks into personally relevant information. Only through this personal meaning of the genetic-information, the information-oriented processes cause inaccurate perceptions. Genetic-counselors are suggested to focus on these personal processes when communicating genetic-information.

1.Introduction

1.1. Inaccuracy of risk perception

Genetic counseling can be described as the 'process of helping people understand and adapt to the medical, psychological and familial implications of genetic contributions to disease' (52). This includes the communication of risk information and medical options based on these risks. These risks are calculated on the basis of a PM DNA-test result, or on the basis of pedigree-information, in combination with the counselees' cancer-history.

On the basis of current literature, we can conclude that genetic-counseling slightly improves the counselees' understanding of cancer-risks. Overall, counselees seem to have a better perception after counseling than before counseling (66,67,78). Yet Smerecnik et al concluded in their review that only approximately 25% (range: 2–55%) of all counselees estimated their risk more correctly after counseling; from an average of 42% pre-counseling to an average of 58% post-counseling. However, on average 25% (range: 5–76%) continued to overestimate and 19.5% (range: 7–55%) continued to underestimate their risk even after counseling (78). Other reviewers concluded that women often have an inaccurate perception when their risk estimates are compared with objective estimate of their risk (77).

Thus, many counselees do not bring their own subjective ideas and feelings about their own cancer-risks in agreement with the actually communicated genetic-information, i.e. the former differs from what has been communicated. Despite being inaccurate in many counselees, the perception of the communicated risks seems to be a better predictor of their medical decisions and distress than the actually communicated risks (277,321,340). Overestimations lead in some individuals to inappropriate uptake of medical surveillance and preventive measures (79,77,203,277,340), and poorer psychological functioning (79,277,321,340).

1.2. Predictors of understanding

Because of the important role of risk-perception and its importance in predicting the impact of genetic-counseling, it is relevant to understand how counselees create their own perception, and especially why it deviates from the actually communicated risks. We differentiate between two kinds of possible causes of the inaccuracy: information-oriented and counselee-oriented predictors. Most previous studies have been information-oriented. Several studies suggest that this orientation should be broadened with counselee-oriented predictors(see 1.4.).

Information-oriented predictors focus on how specific genetic-information is communicated by the genetic-counselor, how it is received, processed and reproduced by the counselee, and how these processes are influenced by knowledge-related variables

such as education and numeracy skills. These predictors focus on the specific processing of specific genetic-information, and not on broad and fundamental processes such as the counselees' personality, and integration of the DNA-test result in their lives in general. In 1.3. we describe the following information-oriented predictors: what information is communicated, levels of education and innumeracy, presence of specific information about cancer in relatives and in counselees, and specific cognitive processes regarding the processing of specific information.

Counselee-oriented predictors focus on how the genetic-information is experienced and fundamentally embedded in the life of the counselee. In 1.4., we describe variables about the self, existential concerns and need for certainty.

Counselee-oriented variables may be important in explaining why counselees have an inaccurate perception of their cancer-risks. Counselees do not perceive their cancer-risks from a cognitive, decontextualised distance, but experience cancer-risks as meaningful for themselves. We suggest that person-oriented predictors about the self, existence and need for certainty influence their risk-perception and its accuracy.

The difference between information-oriented and counselee-oriented predictors could also be described with the difference between the 'function' and 'meaning' of a process. Information-oriented predictors describe how the communication/receipt/inner-processes function in counselees, and the counselee-oriented predictors provide an understanding of the existential meaning of this information for the counselee (cf. difference between the spelling/grammar and the meaning of a sentence). In 1.5. we will hypothesize that counselee-oriented predictors explain (i.e. in statistical terms: 'mediate', cf. 1.5.) *why* information-oriented variables influence the counselees' perception.

1.3. Information-oriented approach: an overview of variables

Presentation format-The format in which cancer-risks are presented by the genetic-counselor may influence the accuracy of the counselees' perception (243,280,281). Genetic risks can be presented as proportions (X out of Y), percentages, and/or in graphical format; risks can be communicated as life-time risks, or related to the current age of the counselee (residual risk, risk over the next 10 years), and can be mirrored (e.g. 80% at-risk implies 20% not-at-risk) (280). Genetic-counselors prefer communication of numerical formats, but few studies provide empirical evidence for its efficacy (279). Explaining general figures of population risks may cause overestimation (278), counselees may feel at fifty-fifty risk (90), and the denominator of proportions are often inaccurately understood (90).Verbal labels or categories are interpreted too subjectively (90).

Communicated information-Cancer-risks may be perceived more accurately when a pathogenic-mutation (PM) is communicated and not an uninformative result (UR) or unclassified-variant (UV), but results are inconsistent (70,86,203,204). There is a large variation in the information communicated during genetic-counseling sessions, which may

influence risk-perception (78). When family-history, heredity and personal risk estimates are all communicated, the counselees' perception of these risks (risk-perception) is more accurate than when only thefamily history, heredity of personal risk is communicated (78). However, we found that counselees may not be able to distinguish the meaning of the DNA-test result from pedigree-information (285), especially after the communication of ambiguous DNA-test results such as unclassified-variants (203).

Question format-In line with the presentation format as described above, also the way how risk-perceptions are measured/formulated by the reseaacher (i.e. the instrument) may influence their accuracy (77). Katapodi et al. (77) concluded in their review that the use of percentage-scales causes larger differences between subjective and objective lifetime risk than Likert-type scales (77). This could be explained by the fact that categorical or Likert-type scales may be more in line with the counselees' own way of describing risks (239). However, percentage-scales may result in the measurement of more accurate perception of 5-years risk (347). The accuracy could be improved by using scales with 7 categories instead of visual analogue scales (348), comparing own risks with general population risks (349,350,348), timing (79) and ordering risk-items in the questionnaire correctly (351). Recent studies suggested that the counselees' medical decisions and distress are better predicted, confirm the counselees' own experiences, when the risk-instruments do not only include cognitive items but also affective items (239), and focus on their interpretations (285,277,203).

Information-related sociodemographics- Lower educated counselees are more unaware of their risks (77), and innumeracy may lead to misunderstanding (79,90,352,353).

Family history-The majority of studies showed that counselees with a positive family history, defined as having at least one first or second degree relative with breast cancer, were more likely to recall/interpret higher cancer-risks than other women, irrespective of their communicated risks (77,354). It has been suggested that family history functions like an 'availability heuristic' (354). Several risk-perception studied included the number/percentage of affected and deceased relatives as predictors. In contrast, the counselee-centered 'lived experience' and personal meaning of being a member of a family with many cancer patients has received little attention (355,328).

Cancer-experience-Affected women seem to interpret their genetic risk in the context of their previous cancer experiences (221), such as recurrence of cancer, surgery and current surveillance. The influence of the counselee-centered meaning of these medical facts on the counselees' perception has hardly been studied.

Cognitive processes-Risk-perception accuracy has also been suggested to be influenced by cognitive information-processes of counselees, such as appraisal, coping and personal theories of inheritance (86,164). Many individuals think in non-Mendelian terms about genetics (354), and use their own rule-of-thumb/heuristics and mental models of inheritance and causes of disease to interpret and assimilate the risk information they

have received (93,94). Counselees have shown to create their own cognitive and emotional representation of the causes, identity, timeline, cure/controllability and consequences of hereditary breast/ovarian cancer (79). These representations may function independently from and/or parallel to rational, factual information (79,95), for instance due to biases of availability, representativeness, anchoring, influence of incidences on risk-perception, emotions and emotional forecasting (90,96). Biases may help people to process information faster (164), cope with health problems, reduce stress (235,298), defend themselves and their self-worth and self-integrity (95). However, the extent to which illness representations in counselees at increased risk differ from healthy individuals may be small (356).

1.4. Counselee-oriented approach

Possibly, previous studies have focused one-sidedly on the communication of probabilities, and have not sufficiently taken into account the personal context and meaning of genetic-counseling for the counselee (38). When confronted with risk-information, counselees have to translate the probabilistic statements into terms with personal meaning (62). 'Taken together (…) risk information is rarely taken up as value-neutral objective truth, but rather risk information is deeply subjective, interiorized against a pre-existing sense of self' (63).

In our previous studies, we developed questions to measure risk-perception that reflected the counselees' own meaning-making process better, instead of risk-perception questions that merely focused on the communication and linear psychological processing of probabilities. We asked counselees about their own interpretations of the meaning of the DNA-test result for their cancer-risks, regardless of what the genetic-counselor has actually communicated (277,285,321,326,340). These interpretations were better predictors of their medical decisions and well-being than their recollections of what the genetic-counselor had actually communicated.

Sense of self: Counselees have to integrate the DNA-test result flexibly in the general life story of who they are (63). They may ask questions about their sense of self such as: Am I a mutation-carrier or not? Am I a potential-cancer-patient or not? And what does this mean? Does this change who I am? Communication of risks may influence/change their identity, and their identity may influence/change risk-perception (61,152). A study in elderly showed that their perception of genetic-risks had been influenced by affect-related personality traits, such as extraversion, optimism, and locus of control (357). Other studies showed that trait-optimism influenced risk-perception (358).

Existence: The counselees' self may be fundamentally involved in risk-perception. Risk-communication may evoke questions about existential concerns in life, such as death, freedom, responsibility, isolation, and meaninglessness (60). Existential feelings may be evoked, such as responsibility for undergoing and disclosing DNA-testing to provide

relatives with risk-information (154-157), guilt about transmitting pathogenic genes to offspring (158), shame and stigma (159,75). As secondary appraisal process, counselees have to integrate, adjust or accommodate the risk-information in their general sense of meaning (131). Counselees have reported that obtaining certainty during the genetic-counseling process had enhanced their lives (6); 83% experienced at least one positive life change (153), and 42% of counselees with unclassified-variant-results reported large changes in their existential view on life (203).

Need for certainty: Counselees do not ask for DNA-testing to understand probabilities accurately (1,5,6), but they want to receive information that provides them with certainty (6,93), e.g. about their own and their relatives' cancer-risks, to know which medical decisions to make (1,5,6,148,149). However, DNA-testing does not provide immediate certainty on demand. Counselees often have to wait for the results for a long time. DNA-test result may be ambiguous, such as UV/URs. Surgery of ovaries and breasts may not be offered after UR/UV. Indeed, counselees have reported that many expectations about genetic-counseling are not met (216,359-361). The counselees' need for certainty often collides with their perceived lack of certainty in the actual situation, causing uncertainty (3,31,164,362-366).

The counselees' need for certainty seem to reflect how the DNA-test result is embedded in their lives. Their unfulfilled need for certainty regarding the DNA-test result, heredity-likelihood and cancer may influence the way how they perceive cancer-risks. Counselees who do not receive certain genetic-information may re-interpret this information in such ways that they do perceive certainty. Therefore, many counselees seem to attach more value to their own opinion than to the genetic-counselor's (285,203).

These counselee-oriented processes may also explain why the information-oriented processes influence their perception. That is, the presentation and question format, the communicated information, sociodemographics and family history may influence the counselees' perception, because of the meaning of this information for their selves, their existence and fulfillment of their needs for certainty. The counselee-oriented processes may motivate them to use cognitive techniques; for instance, counselees with a large need for certainty but who perceived uncertainty over the DNA-test result may distort the information in their perception to perceive certainty. In summary, we expect that counselee-oriented variables predict the accuracy of their perception with equal or larger effect sizes than information-oriented variables, and completely mediate the effects of information-oriented variables on this accuracy.

1.5. Research questions

1. Do counselee-oriented variables regarding the self, existence and the unfulfilled need for certainty significantly predict the accuracy of the counselees' perception?
2. Is perception-accuracy also significantly predicted by information-oriented predictors?

3. Do counselee-oriented variables significantly explain the effect of information-oriented variables on the accuracy of the counselees' perception; more specifically: when counselee-oriented variables are included in the analyses, do the effects of the information-oriented variables on the accuracy of the counselees' risk-perception become non-significant?

2. Method

2.1. Procedure and design

Eligible participants were women with breast and/or ovarian cancer who had requested for a BRCA1/2-test in the period 2006-2009 at the departments of Clinical Genetics of the Leiden University Medical Center, the Maastricht University Medical Center, the University Medical Central Groningen, Erasmus Medical Center Rotterdam, or the VU Medical Center Amsterdam. Eligible counselees received two questionnaires: immediately after the first genetic-counseling session (T1), and 3 months after the second genetic-counseling session in which the DNA-test result was disclosed (T2). Usually, genetic-counselors disclosed the following information: DNA-test result category, heredity-likelihood, cancer-risks for female relatives and for the counselee, risk management options (surgery, surveillance) for relatives and counselees, including the possibility for relatives to undergo DNA-testing when applicable. Table 3 in chapter 6 shows the most frequently communicated information; more details on procedure, design and population are described elsewhere (340).

2.2. Instruments

All instruments are presented for T2 only. Using counselee-oriented and/or information-oriented predictors at T1 yielded similar results (not presented).

The accuracy of the counselees' risk-perception was measured as difference between the counselees' interpretation of their own cancer-risks and the cancer-risks actually communicated by the genetic-counselor. We decided to use their interpretations and not their recollections of cancer-risks (285), because previous analyses in the same sample showed that their interpretations did not differ significantly from and correlated strongly with their recollections (340), but did predict psychological and medical outcomes better than recollections (340,277). Interpreted cancer-risks were measured by the question 'regardless of what your genetic-counselor has communicated, what are your own thoughts and ideas of your risks to develop cancer?' Counselees could answer on a 1-7 Likert-scale ranging from 1 (not at risk) to 7 (complete at risk), which had the least number of missing values compared with percentage scales and showed the most accurate perception(285,277). Actually communicated risks had been derived from a checklist filled-in after each session by the genetic-counselor, medical-files and summary letters that

counselees received within 3 months after the DNA-result; actual risks were rescaled to the 1-7 Likert-scale to match the counselees' interpretation. We used absolute-difference scores (i.e. regardless of the direction of the difference), because of the explorative nature of the study, the small sample size and missing values; moreover, unpresented data analyses did not show different results when we did not use absolute differences.

Items about the self were measured by the Ryff-well-being questionnaire, which was shown to be a reliable and valid scale to measure positive, existential well-being; we used the scales: autonomy, mastery, vitality, inner strength, and self-acceptance (319,367,368). In addition, we used the Revised Life Optimism Scale to measure trait optimism, which was shown to be a reliable, valid instrument (320,369).

Existential items were measured with the purpose-in-life-scale of the Ryff well-being scale (319,367,368). Further, we asked counselees to rate on three 1-7 Likert-scales (1, very seldom, 7-very often): how often they had been wondering how many years they still have to live, what the meaning of their life is, and how often they actually experienced their life as meaningful during the last two weeks.

Need for certainty was measured with the Need for Structure Scale, which is a reliable, valid instrument to measure one's desire for structure and response to lack of structure (370). We asked them about the number of experiences with uncertainties in life before genetic-counseling, and the number of certainties; answers were given on 1-7 semantic differentials (1, little experiences, 7, many experiences). In chapter 10, we describe how we developed items about the Unfulfilled Need for Certainty regarding the domains of DNA-test result, heredity, cancer and self (371). We asked counselees to rate on 7-point scales to what extent they wished to receive certainty about these domains; from this need for certainty we subtracted the level of certainty that they perceived during last two weeks.

Information-oriented variables were developed on the basis of literature and of the experience of involved genetic-counselors. Table 2 describes instruments for presentation format, communicated information, sociodemographics, family history, cancer-experience, cognitive processes. Specific coping styles were measured with the COPE (318) regarding coping with the DNA-test result. Regarding the question-format we only present Likert-scales in this article. We have also asked counselees to recall and interpret their risks in percentage-scales and we compared this with actually communicated cancer-risks; this did not result in different study outcomes.

Table 2. *Overview of information-oriented instruments*

Information-oriented group of variables	Variable description	Items/scales
Presentation format		**Risks communicated in words**, graphics, percentage, proportion or **in a combination of formats**; **mirroring of risks**; exact cancer-risk versus range of cancer-risks; using the term 'genetic change' or using other terms (all binary items)*
Communicated information	DNA-test result category	**Pathogenic-mutation; uninformative-result (i.e. no mutation is found, but counselee is at risk because of pedigree); unclassified-variant (i.e. mutation is found for which the pathogenic meaning is not know yet, and counselee is at risk because of pedigree).**(all binary items) *;**
	Cancer-risks and heredity	**Cancer-risks for proband(%); cancer-risks for relatives(%);** likelihood that cancer is heritable in the family (heredity-likelihood; verbal) *; ** (285)
	Additional counseling aspects	**Counseling was face-to-face; a flyer explaining genetic-testing/results was provided. During the intake: possibility of finding an unclassified-variant mentioned**; explanation of population breast/ovarian cancer-risks; explanation of part of breast/ovarian cancers caused by heredity; risk of finding a pathogenic-mutation; risk of transmitting a pathogenic-mutation when detected. Communicated during result-disclosure: additional explanation of the detected mutation; mutations –also benign ones- are frequently found in DNA; being-at-risk does not mean developing cancer; cancer is not likely to be heritable in your family; other untested mutations may explain cancer; extra explanation of genetics in general; (im)possibilities of DNA-testing; start of family-research of DNA-test result in relatives; possibility of future research and new findings. (all binary items, except for risks measured in %) *
Knowledge-related sociodemographics		educational level (both measured binary, i.e. higher/lower than high school, and on 7-points scale) ***
Family history		Pedigree-information: high cancer-risk; moderate cancer-risk; low cancer-risk *;**
Cancer experience	Medical history	Binary items: breast cancer, ovarian cancer, metastases, preventive bilateral mastectomy (PBM), bilateral salpingo-oophorectomy (PBSO), chemotherapy, radiotherapy, other therapy; elapsed years since cancer diagnoses, metastases, treatment and DNA-test result***
Cognitive processes	Recollections and expectations	Measured at intake: 'Before genetic-counseling, to what extent did you expect to receive a pathogenic-mutation'; measured after result-disclosure: to what extent was this DNA-test result in line with your expectations; **do you expect to receive a new DNA-test result in the future; what extent of heredity-likelihood do you expect this future result to imply?****
	Illness representation	Influence on life; **duration**; control; helpfulness of treatment; **severity/physical limitations**; worries; understanding; influence on mood (semantic differentials, 0, not, 10, completely)***(372)
	Coping	Scales of COPE: **Active, acceptance, priority-taking, planning, renaming,** denial, distraction, turn to God, waiting, taking drugs. Scale of IES: **avoidance*****(318,286)

*Instrument: *Filled-in by genetic-counselors in a checklist after each genetic-counseling session; **derived from medical-file and/or summary letter sent to the counselee by the genetic-counselor; ***counselees' questionnaire. Social variables are not included (see discussion-section); **Bold:** significant predictors of the inaccuracy of perception (see table 3)*

2.3. Statistics

Missing values were imputed by multiple imputing. Population variables are described with frequencies and means.

Question 1: We performed regression-analyses with one counselee-oriented predictor (X) at a time (standardized ß=R) to predict the perception-accuracy (Y); due to multicollinearity and the relatively small sample, we could not use multiple-predictor-analyses.

Question 2: We did the same for information-oriented variables (X). Differences in X and Y between DNA-test results were tested with Kruskal-Wallis tests (K-W); DNA-test results were included as moderators in regression analyses to test differences in the relationships between X and Y; only significant differences between DNA-test results are presented in this chapter, and otherwise we show overall results.

Question 3: We did mediation analyses (184,185) as we described elsewhere (277,340). We did one analysis for each information-oriented variable. The information-oriented variable was the predictor (X), the predicted variable was the perception-accuracy (Y). Mediators were the counselee-oriented variables (M). Mediation is assumed to be present when four criteria are met. 1. X and M correlate. 2. X predicts Y. 3. M predicts Y. 4. When both X and M are included in prediction of Y, and we compare these results with criterion 2, the predictive value of X decreases (i.e. partial mediation) or becomes non-significant (i.e. complete mediation). For example, the communication of a UV-result instead of PM/UR-relates correlates with a strong unfulfilled need for certainty about the DNA-test result (step 1); both the UV-result and the unfulfilled need for certainty predict a more inaccurate perception (steps 2 and 3); when the unfulfilled need for certainty is included in analyses, the UV-result does not significantly predict the accuracy anymore which suggests complete mediation (step 4).

We did multiple mediation analyses.

Firstly, we did mediation analyses with all counselee-oriented variables together as mediators, but for some information-oriented variables this resulted in a very small number of participants per analysis due to missing values or multicollinearity.

Secondly, we did mediation analyses with only autonomy, purpose in life and unfulfilled need for certainty about the DNA-test result together as mediators; these variables correlated strongest with both the significant information-oriented predictors and the accuracy; the number of participants per cells was large enough to calculate this.

Third, we did analysis for each of these three counselee-oriented variables separately, to make the number of participants in each analysis as large as possible. For presentation purpose, we only present data with p-values<.01 and std.ß>.20, and only show tables for mediation criteria 2 and 3. Criterion 4 is not presented because we only found complete mediation. Significance level was defined as p<.01. This level reflected a balance between

the explorative nature of this study (suggesting to set a high p-value to avoid type-II error), and the large number of tests (suggesting a low p-value to avoid type-I error).

3. Results

3.1. Population

We approached 654 women who had undergone BRCA1/2-testing. Of them, 467(71%) filled-in the T1-questionnaire and 248(53%) the T2-questionnaire. Mean time since cancer-diagnosis was 5 years; 94% had had breast cancer and 6% ovarian cancer. Metastases were present in 26% of all participants. Before DNA-testing, 56% had undergone symptomatic mastectomy, 6% symptomatic bilateral salpingo oophorectomy (BSO) and 5% preventive/presymptomatic (PBSO). Mean age was 56 years, 42% had visited high school or higher, 84% was married, 87% had children. More information is published elsewhere (340). Missing value-analyses did not show significant results.

3.2.Counselee-oriented predictors

Self: Counselees had a more accurate perception when they were more autonomous, felt more mastery, vitality, self-acceptance, optimism, and inner strength. Effect sizes were moderate to large (see table 3).

Existence: Counselees perceived their cancer-risks as more accurate when they had a stronger experience of purpose in life, less frequently wondered how many years they still can live and what the meaning in their life is, and currently experienced living a more meaningful life. Effect sizes were moderate to large.

Need for certainty: Counselees perceived their cancer-risks as more accurate when they were more experienced with uncertainties in life, desired less structure and reacted more positively to a lack of structure. Accuracy was also higher when counselees reported less needs for certainty, perceived more certainty and experienced more fulfillment of their needs for certainty about cancer, DNA-test result and heredity. Effect sizes were moderate to large.

Table 3. *Results for counselee-oriented predictors*

	Counselee-oriented predictors	Inaccuracy of perception (std.ß)
Self	-Autonomy	-.24
	-Mastery	-.35
	-Vitality	-.24
	-Self-acceptance	-.23
	-Optimism	-.28
	-Inner strength	-.20
Existence	-Purpose in life	-.20
	-Wondering about how many remaining years	.35
	-Wondering about meaning in life	.43
	-Experiencing meaning in life	-.39
Need for certainty	-Desire for structure	.39
	-Reaction to lack of structure	.39
	-Previous experiences with uncertainties in life	-.25
	-Need for certainty about DNA-test result	.20*
	-Need for certainty about heredity	.48
	-Need for certainty about cancer	.22*
	-Need for certainty about the self	.20*
	-Perceived certainty about DNA-test result	-.34
	-Perceived certainty about heredity	-.40
	-Perceived certainty about cancer	-.40
	-Perceived certainty about the self	-.35
	-Unfulfilled need for certainty about cancer	.40
	-Unfulfilled need for certainty about DNA-test result	.36
	-Unfulfilled need for certainty about heredity	.55
	-Unfulfilled need for certainty about the self	.39

*All p<.01, std.ß>.20; *p<.07; results regard the total sample because no significant differences were found between DNA-test results (pathogenic-mutation; uninformative-result; unclassified-variant; not-significant Kruskal-Wallis tests); a positive ß means that the counselee-oriented predictor had caused a more inaccurate perception, and a negative ß means a more accurate perception.*

3.3. Information-oriented predictors

Table 4 presents the significant information-oriented predictors (P<.01; std.ß>.20). About half of the tested information-oriented variables were significant predictors (cf. table 2). All significant effects were moderate to large.

Presentation format: The counselees' perception was more accurate when: risks for UV/URs had not been communicated in words or in multiple formats. The perception was also more accurate when the cancer-risks for UV/URs had not been mirrored (e.g. 80% at risk = 20% not at risk), and when these risks for PM results had been mirrored.

Communicated information: The counselees' perception was more accurate when a PM and not an UR or UV had been communicated, when cancer-risks were higher, when counseling was face-to-face and a flyer had been provided, and when the possibility of finding an unclassified-variant had been mentioned during the intake.

Cognitive processes: The higher the cancer-risks and heredity-likelihood were in the counselee's recollections and/or expectations, the more accurate was their risk-perception. The perception was more accurate when counselees expected that the duration of their cancer would be shorter, cancer was less severe, and when they used active, accepting, priority-taking and planning coping styles, and did not use renaming or avoidance as coping.

3.4. Mediation analyses

When we included counselee-oriented variables as mediators (M) in regression-analyses, information-oriented variables (X) did not significantly predict the level of accuracy of the counselees' perception anymore (Y), and the counselee-oriented variables were the only significant predictors of Y (cf. table 3). Information-oriented variables became non-significant when we used them as mediators: all counselee-oriented variables, the three strongest variables (autonomy, purpose, unfulfilled need for certainty about DNA-test result) and each of these variables separately.

Table 4. *Results for information-oriented predictors*

Information-oriented predictors		Inaccuracy of perception (std.ß)
Presented format	Counseling format: in words	.29(UV/UR)/ns(PM)
	Counseling format: combination of formats	.30 (UV/UR)/ns(PM)
	Mirroring of risks	.40 (UV/UR)/-.58(PM)
Communicated information	Actual pathogenic-mutation	-.23
	Actual uninformative-result	.24
	Actual unclassified-variant	.20
	Actual cancer-risks proband	-.38
	Actual cancer-risks relatives	-.30
	Face-to-face counseling	-.20
	Provision of flyer explaining genetic-counseling	-.33
	Possibility of finding an unclassified-variant mentioned during the intake	-.42
Cognitive processes	Recalled own cancer-risk (1-7 Likertscale)	-.35
	Recalled own cancer-risk (% scale)	-.35
	Recalled relatives' risk (1-7 Likertscale)	-.46
	Recalled heredity-likelihood (1-7 Likertscale)	-.34
	Expectation of a new result in future	.40
	Expectation of a pathogenic-result in future	.47
	Expected duration of cancer	.33
	Experienced physical symptoms/severity of cancer	.35
	Active coping with DNA-test result	-.41
	Acceptance coping with DNA-test result	-.28
	Priority coping with DNA-test result	-.39
	Planning coping with DNA-test result	-.34
	Renaming coping with DNA-test result	.38
	Avoidance coping with DNA-test result	.29

P<.01; std.ß>.20 Non-significant results not presented; T=t-test is significant (p<.01) with medium or large effect; N=n<50; ns=not significant; results regard the total sample, except were reported, because no significant differences were found between DNA-test results (pathogenic-mutation (PM); uninformative-result (UR); unclassified-variant(UV); not-significant Kruskal-Wallis tests); a positive ß means that the counselee-oriented predictor had caused a more inaccurate perception, and a negative ß means a more accurate perception.

4. Discussion

4.1. Conclusions

This study showed that both counselee-oriented and information-oriented variables predicted the accuracy of the counselees' risk-perception. The amounts of variance explained by counselee-oriented and by information-oriented variables were similar (cf. tables 3 and 4).

We found that several information-oriented variables influenced the accuracy of the counselees' risk-perception. Counselees with UV/UR's were less accurate than PM-carriers, possibly because this information was less clear; communication of additional information in words and/or in multiple formats seemed to have confused these counselees even more (cf. 203). PM-carriers and counselees with high risks (actually communicated and recalled) had a more accurate perception, possibly due to the clarity of pathogenic results which may be easier to perceive accurately. As expected, their perception was more accurate when counselees were younger (77), counseling was face-to-face, and flyers were provided (327). Moreover, counselees had a more accurate perception when they had active coping styles, and did not have passive coping styles, negative expectations and distress, which confirms other studies (90,284).

The influence of information-oriented variables was completely mediated/explained by counselee-oriented variables. We found that the counselees' risk-perception was directly influenced, and was completely mediated, by the following variables: positive-existential personality characteristics, experience of meaning/purpose in life, previous experiences with uncertainties in life, their general need for structure, and their specific needs for certainty about the DNA-test result, about heredity of cancer in their family, and about their own cancer. These counselee-oriented variables completely explained the effect of the information-oriented variables.

The information-oriented variables influenced the counselees' perception *because* they evoked a personal process in the counselee, which involved her self/personality, her existential concerns in life and her needs regarding the DNA-test result, her cancer and the heredity of cancer in the family. For instance, the presentation format and the actually communicated DNA-test result have influenced the counselees' risk-perception, *only via* (i.e. through the mediation of) the personal and existential meaning that this information inherently has for the counselees. More specifically, the communication of a PM (i.e. information-oriented) created a feeling of certainty over the genetic cause of cancer in the counselee (i.e. counselee-oriented); subsequently this feeling of certainty influenced the counselees' perception, and created an *indirect* relationship between the communication of the PM and the perception.

The results also suggest that the counselees' cognitions, such as their cognitive illness representations, influence their risk-perception *through* the personal and existential

meaning of these cognitions. Thus, the counselees' risk-perception is not determined by merely rationally knowing 'I am at risk', but by the personal and existential meaning of knowing this. Thus, when counselees are confronted with risk-information, they translate the probabilistic statements into terms with personal meaning (62), and try to embed this information in the general story of their lives (59). By subjectively translating and embedding this information, counselees seem to distort the originally communicated cancer-risks, i.e. creating their inaccurate perception.

4.2. Counselee-oriented approach

The counselee-oriented predictors that we propose in this chapter are not intended to replace the information-oriented predictors. Our approach is integrative and is intended to understand/explain how information-oriented processes do influence the accuracy of the counselees' risk-perception. First, we described the impact of actually communicated genetic-information on the counselees' perception, and described how counselees function psychologically, such as using biases and heuristics. Subsequently, we explained by mediation analyses why counselees experienced these information-oriented influences.

We suggest that the perception of cancer-risks is not a sum of 'decontextualized' (38) representations, biases, rules or schemas. Risk-perception is the result of a counselee putting the communicated risks in the lived experience and broad context of her life, which includes how she manages existential concerns and needs, and what kind of person she is. Because of her fundamental needs, she may use cognitive techniques and misinterpret the communicated risks.

The counselee-oriented approach seems to be a less normative approach than the information-oriented approach. To be in line with other studies, we used the term 'inaccurate perception'. However, words such as 'inaccuracy' and 'inadequate counseling' seem to suggest that the counselee and/or genetic-counselor are 'wrong'. But even if a counselee may be 'inaccurate' from an information-oriented point-of-view, she could feel justified from her own point-of-view and from her own needs and drives. Thus, having an inaccurate perception may not necessarily mean that counselees want to be provided with additional 'correct information'. It could be argued that letting counselees have their own inaccurate perception – i.e. respecting their autonomy - may sometimes be more ethically justified than paternalistically forcing them 'to think adequately'.

The counselee-oriented approach is in line with the general trend in psycho-oncology to pay more attention to the role of meaning and spirituality in cancer-patients (122). For instance, cancer-patients who experience meaning in life seem to be better adjusted to cancer (132,144,146,147). They also experience a better quality-of-life and well-being, and up to 50% less depression (132,133,139-141).Terminal patients with high spiritual well-being also experience a lower desire for hastened death, less depression and less suicidal ideation (133,142). Moreover, patients who are able to reengage in meaningful

goals and focus on pleasant issues, experience more positive affect (143). These meaning-making processes may be influenced by the way that physicians communicate with patients (373,374).

4.3. Limitations and implications

Our study is based on a relatively small sample size in the moderately short follow-up time after DNA-test result disclosure. The study may be biased by the fact that we have only included women affected with cancer. However, elements in their cancer-history –such as having metastases- did not predict the level of accuracy of the counselees' perception, and were also not-significant as moderators in unpresented interaction analyses. We suggest replicating this study in other genetic disorders, in both sexes and in patients affected and unaffected with cancer.

Subsequent studies should also focus on the social construction of the meaning of genetic-information, because friends and relatives are part of the counselees' context, and may influence their interpretations (90,375,376). Such social studies should not focus on the open communication of genetic-information per se (168), but on the experience and the meaning of this communication for the counselee, such as perceived social support. More counselee-oriented instruments could be developed and used. For instance, interview studies could be performed to ask counselees what the communicated information fundamentally means to them, like in before-mentioned qualitative studies. Moreover, future studies may also examine which variables (dynamically) limit the counselee from sharing her subjective ideas and feelings in an information driven counseling session.

Regression mediation analyses strongly indicated the presence of mediation effects, but did not definitely prove this, because this was not an intervention study (188). However, the design of the study and the nature of the mediators made mediation likely (188). These mediation results confirmed our theoretical expectations and previous qualitative studies. T2-accuracy was predicted by T1-predictors. The communication of DNA-test results did actually influence the mediators like an intervention (as described elsewhere, i.e. 371). Questionnaires/items about the self also showed to be flexible instead of being an unchangeable trait which would suggest moderation instead of mediation; because we found significant differences between T1 and T2 in the self-items (d>.4), suggesting that the DNA-test result may have altered the counselees' self-experience which is in line with our hypotheses (60,cf.61-62). We suggest performing intervention studies to determine and influence these mediation effects.

From our counselee-oriented perspective, we suggest to focus on developing assessment instruments and interventions regarding the psychological/existential needs and motivations of counselees to undergo genetic-counseling. Genetic-counselors could

explicitly ask counselees about their reasons to request DNA-testing, and assess the role of this request in their actual life situation.

Subsequently, genetic-counselors could use this counselee-oriented assessment to explore together with counselees the decision to undergo DNA-testing or not. DNA-testing may not be suitable for all counselees in all personal/existential situations, and some counselees may first need psychological counseling. Different stages of readiness to undergo DNA-testing may exist (377). For instance, does a counselee have (too) high expectations of DNA-testing as a way to cope with her cancer? Could some of them benefit more from first referring them for intensive medical and/or psychological help to learn to live with cancer, instead of immediately undergoing DNA-testing? When counselees receive counseling at an optimal stage, their perception of genetic-information and its consequences may be better adjusted to the actual medical situation. Current theories and instruments about stages-of-readiness should be developed to include counselee-oriented elements.

The counselee-oriented assessment could also be used in tailoring the format of risk-communication, for instance by focusing on the personal consequences of the DNA-test result (38). Such assessments before DNA-testing could include the question how counselees think and feel about their cancer-risks; the actual risks could be tailored to this pre-testing risk-perception. Such interventions seem to make the counselees' risk-perception more accurate (282). To assess their understanding after DNA-test result disclosure, counselees could be asked to repeat the communicated information in their own words. To explore their interpretations, counselees could be asked about 'their own ideas and feelings, regardless of the communicated information' and about the medical consequences they have in mind (277,285,340).

Counselees could be provided with additional information if necessary. But as our study suggests, information-provision alone cannot be expected to improve their perception much. Therefore tailored risk-communication may also include discussion of the fundamental subjective meaning that the DNA-test result may have for counselees, and the ways how they can embed the result in their lives. For instance, a counselee could explore together with her genetic-counselor what she can do with this risk-information, what she can tell their relatives, and how this information feels, and how she copes with uncertainty and vulnerability related to this result.

To help counselees in creating a realistic meaning of the DNA-test result, it may be explained beforehand what counselees can realistically expect from genetic-counseling and what not. This regards both medical and psychological aspects, including both certainties and uncertainties. Currently, patient-information is often unbalanced in the Netherlands, because flyers and websites seem to pay much attention to certainties and little to uncertainties that may arise after DNA-testing. Nuanced patient-information could

help counselees to have realistic expectations of the certainties they may obtain, which may subsequently improve their perception of the communicated information.

More rigorous interventions to improve the counselees-oriented variables include training in coping with hereditary cancer (331), and (continue) finding meaning in life despite one's cancer-experience, physical limitations and (genetic) uncertainties (378). Several existential psychotherapeutic interventions for cancer-patients have been developed, showing moderate effects on distress and well-being (138,379); these effect sizes are comparable with other psychological interventions for cancer-patients (380-382). More recent meaning-oriented interventions have shown to have large effects (378,383,384).

Our advice to develop counselee-oriented interventions is in line with the review of Edwards et al (327). They showed that previous interventions, both information-oriented and counselee-oriented, have not been effective because of their information-oriented elements, but because of their counselee-oriented elements, i.e. focus on emotions and support (327). It has been advised to develop genetic-counseling into a personal, two-directional/reciprocal process (283) with explicit focus on the counselee. This may help counselees in their search for certainty, may improve their perception of genetic-information and make their medical decisions more well-informed.